AN EVALUATION OF 820 ORBITAL CASES*

BY *Robert E. Kennedy,* MD

INTRODUCTION

ORBITAL PROBLEMS CAN BE OF GREAT CONCERN TO THE PATIENT, FAMILY, AND physician. Such problems may be developmental, inflammatory, associated with general or metabolic disease, traumatic, either benign or malignant, neoplastic, vascular, or of unknown etiology. For the patient there can be permanent cosmetic deficit, ophthalmological deficit with loss of vision or the eye, and even loss of life. Medically, orbital problems can be a challenge in diagnosis and management which may require a team approach to include the ophthalmologist, ophthalmic surgeon, internist, otolaryngologist, neurosurgeon, plastic surgeon, radiation therapist, and oncologist.

While the spectrum of orbital problems in an ophthalmic practice is broad, the reported incidence of orbital mass lesions varies considerably depending on the origin of the report as reviewed by Jones and associates.[1] These reports differ substantially depending on whether they are clinical,[2,3] radiologic,[4] from pediatric centers,[5-10] neurosurgical departments,[11,12] pathology referral centers,[13-16] clinical referral centers,[17-19] or from different geographic areas.[20]

There is a significant difference in the series determined from biopsy only and those which include clinical diagnoses without biopsies. While the former provide more exact figures on which to base prognostic information, the latter are more representative of what would be anticipated in the average ophthalmic practice.

MATERIALS AND METHODS

The current study was made to determine the frequency of various orbital problems as encountered by the general practicing ophthalmologist in an average community which is not a large referral center. Rochester is a community with almost a quarter million population in Upstate New

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York. The surrounding county raises the total to about 700,000. There are approximately 60 ophthalmologists in this area.

Based on numerous inquiries, the average ophthalmologist in this area sees a bonafide case about once every 3 to 5 years and invariably refers the patient. Most ophthalmologists average two to three endocrine thyroid patients per year. These are rarely referred unless surgery is indicated. This study includes: (1) orbital biopsy proven lesions, (2) orbital cases without biopsy when there was very strong medical, x-ray, or computerized tomography (CT) evidence, positive biopsy of other body lesions, or known metastasis, and (3) direct extension to the orbit from lid and conjunctival tissue. Excluded from this study are lesions (1) confined to the eyelids and conjunctiva, (2) intraocular, and (3) Grave's endocrine disease unless operated for possible tumor, corneal exposure, optic nerve deficit, or requiring orbital decompression.

In addition, traumatic orbital problems are included. The data represent patients who have been seen at six hospitals in the community, referred by local ophthalmologists, or from surrounding towns or nearby cities. These patients have all been seen since September 1949 and managed by me personally or in conjunction with the referring doctor. I owe a debt of thanks to most of my colleagues for their confidence, cooperation, and trust in referring these patients for evaluation and disposition over a 34-year period.

Eight hundred twenty orbital cases were reviewed during the last 34 years. These are classified in general categories in Table I, together with the number in each category and percentage of the total of 820 cases. In Table II the categories are further broken down as to the number of various types of involvement in each category. Grave's or thyroid orbital involvement is limited to those cases which were operated on. These categories can be used as a clinical classification of orbital problems.

DISCUSSION

The advantages of biopsy-proven series are recognized for the certainty and prognostic aspects. However, such series may not include accurate figures, for many virtually certain clinical lesions are usually not biopsied, such as hemangiomas. This series includes many such cases which are almost certainly diagnosed based upon medical findings, x-rays, CT, or a biopsy from another site. These are meaningful in a clinical series (Figs 1 to 3). Other similar cases are included.

How accurate is an assumption that the unbiopsied orbital lesion would be the same as that of another known primary site? In this series, one

TABLE I: CLASSIFICATION OF ORBITAL CASES

	NO	%
Vascular	72	8.8
Lymphoid tumors/leukemia	106	13.0
Orbital inflammations	91	11.1
Grave's disease (surgery only)	47	5.7
Lacrimal gland	45	5.5
Neurogenic tumors	68	8.3
Rhabdomyosarcomas	8	1.0
Secondary orbital tumors	51	6.2
Metastatic	27	3.3
Congenital and developmental	13	1.6
Pseudotumor/granuloma	43	5.2
Cystoid tumor	89	10.9
Mesenchymal tumors	7	0.9
Fibroosseous tumors	26	3.2
Trauma	107	13.0
Normal orbital tissue	2	0.2
Undiagnosed	18	2.2
Total	820	100

female, age 68, had a breast carcinoma 5 years earlier and presented with a left orbital mass. Orbital biopsy proved this to be a lymphosarcoma, a different tumor than what might have been expected, with an occurrence rate of less than 0.15%. The statistics are altered very little if biopsy is not accomplished and much undesired surgery is avoided.

Traumatic orbital problems are also included in this clinical series and are capable of leaving marked deficits or even be life-threatening (Figs 4 to 6). Many fractures,[21] transorbital stab wounds, intraorbital foreign bodies, and gunshot wounds, although appearing trivial, may have underlying serious intracranial involvement requiring close team attention including the ophthalmologist, otolaryngologist, neurosurgeon, and internist. The incidence of intracranial involvement can be as high as 25%.

The following case illustrates the dilemma which can be encountered by the clinician treating orbital cases, but who is somewhat isolated by not practicing in a large medical center. A 28-year-old male noted visual distortion in his right eye and was found to have low-grade proptosis and retinal striae with ultrasound and x-ray evidence of an orbital tumor. An orbital exploration was performed temporally (Fig 7). Pathologic readings were obtained from various departments and included:

1. Hemangioendothelioma
2. Hemangiopericytoma
   Question of origin in lacrimal gland
   Question of invasion into lacrimal gland

3. Pleomorphic adenoma, low malignancy
4. Fibrous histiocytoma
5. Cellular neurofibroma
6. Neurofibromatous sarcoma, low malignancy

Recommendations for management were also varied and included:

1. Firm reassurance
2. Reassurance with yearly examinations and CT scan studies
3. A course of radiation therapy
4. Chemotherapy
5. Radiation and chemotherapy
6. Exenteration

Since there is a wide spectrum in management between pleasant reassurance and exenteration, even after several pathologic opinions, this leaves the clinician in an awkward position in recommending the best management.

With the thought that the lesion might well be a hemangiopericytoma it brings to our attention the previous reports by Jakobiec et al[22] and Henderson and Farrow[18] that in a small series this tumor can require exenteration (Jakobiec, 2 of 7 cases) and can be significantly life-threatening (Henderson, 3 of 11 cases) with a high incidence of recurrence (Henderson, 2 of surviving 8 cases). This aggressive behavior and potentially lethal nature of some hemangiopericytomas primary to the orbit should be known to the orbital surgeon for advice to the patient.

TABLE II: BREAKDOWN CLASSIFICATION OF ORBITAL CASES

	NO		%
Vascular		72	8.8
Capillary hemangioma	22		
Cavernous hemangioma	14		
Hemangiopericytoma	7		
Hemangioendothelioma	2		
Lymphangioma	4		
Carotid-caverous fistula	9		
Cavernous sinus thrombosis	3		
Varices	6		
Miscellaneous	5		
Surge-Weber syndrome	1		
Lymphoid tumors and leukemia		106	13.0
Lymphoma	56		
Lymphosarcoma	31		
Burkitt's lymphoma	2		
Reactive lymphoid hyperplasia	3		
Myelogenous leukemia	6		
Multiple myeloma	2		
Hodgkin's disease	2		

TABLE II: BREAKDOWN CLASSIFICATION OF ORBITAL CASES
(Continued)

	NO		%
Eosinophilic granuloma	2		
Histiocytosis X	1		
Reticulum cell sarcoma	1		
Orbital inflammations		91	11.1
Idiopathic inflammatory pseudotumor	43		
Dacryocystitis	2		
Vasculitis	2		
Wegener's granulomatosis	2		
Giant cell reparative granuloma	1		
Sarcoidosis	3		
Orbital cellulitis/abscess sinus	32		
Osteomyelitis-fistula	6		
Grave's disease [thyroid] (surgery only)		47	5.7
Decompression	21		
Vision suppression	10		
Isolated myositis	5		
Pseudotumor *vs* myositis	11		
Lacrimal gland		45	5.5
Carcinomas	5		
Adenocarcinoma (2)			
Adenoid cystic (3)			
Mixed cell	12		
Dacryoadenitis	12		
Lymphocytic infiltration	8		
Cyst	4		
Lymphoid hyperplasia	4		
Neurogenic tumors		68	8.3
Neurofibromatosis	25		
Schwannoma	1		
Neurolemoma	3		
Meningioma	24		
Arachnoid hyperplasia	1		
Optic nerve gliomas	8		
Astrocytoma	2		
Ependymoma	1		
Pituitary	3		
Rhabdomyosarcoma	8	8	1.0
Secondary orbital tumors		51	6.2
Retinoblastomas	10		
Melanomas	4		
Malignant sinus extension	18		
Conjunctival squamous carcinoma	3		
Lid basal cell extension or squamous cell	14		
Melanoma of lid	1		
Salivary gland extension	1		

TABLE II: BREAKDOWN CLASSIFICATION OF ORBITAL CASES
(Continued)

	NO		%
Metastatic		27	3.3
Neuroblastoma	2		
Breast	8		
Lung	3		
Kidney	1		
Prostate	4		
Melanoma	2		
Undetermined	7		
Congenital and developmental		13	1.6
Craniostenosis facial asymmetry Hypertelorism Crouzons	11		
Facial cleft	2		
Pseudotumor/granuloma	43	43	5.2
Cystic tumors		89	10.9
Dermoids	45		
Mucocele	31		
Meningocele	1		
Encephalocele	2		
Epithelial cysts	8		
Blood cyst orbit	2		
Mesenchymal and fibroosseous tumors		7	0.9
Fibrous histiocytoma	1		
Chondrosarcoma	1		
Malignant mesenchymoma	1		
Lymphoepithelioma	1		
Postirradiation osteogenic sarcoma	1		
Fibroantioma	2		
Fibroosseous tumors		26	3.2
Osteoma	6		
Ossifying fibroma	2		
Fibrous dysplasia	9		
Paget's disease	8		
Fibroosteoma	1		
Trauma		107	13.0
Weapon bullet	7		
Orbital hematoma	3		
Foreign body in orbit	12		
Stab	4		
Miscellaneous	3		
Fractures	78		
Undiagnosed	18	18	2.2
Normal orbital tissue	2	2	0.2
Total		820	100

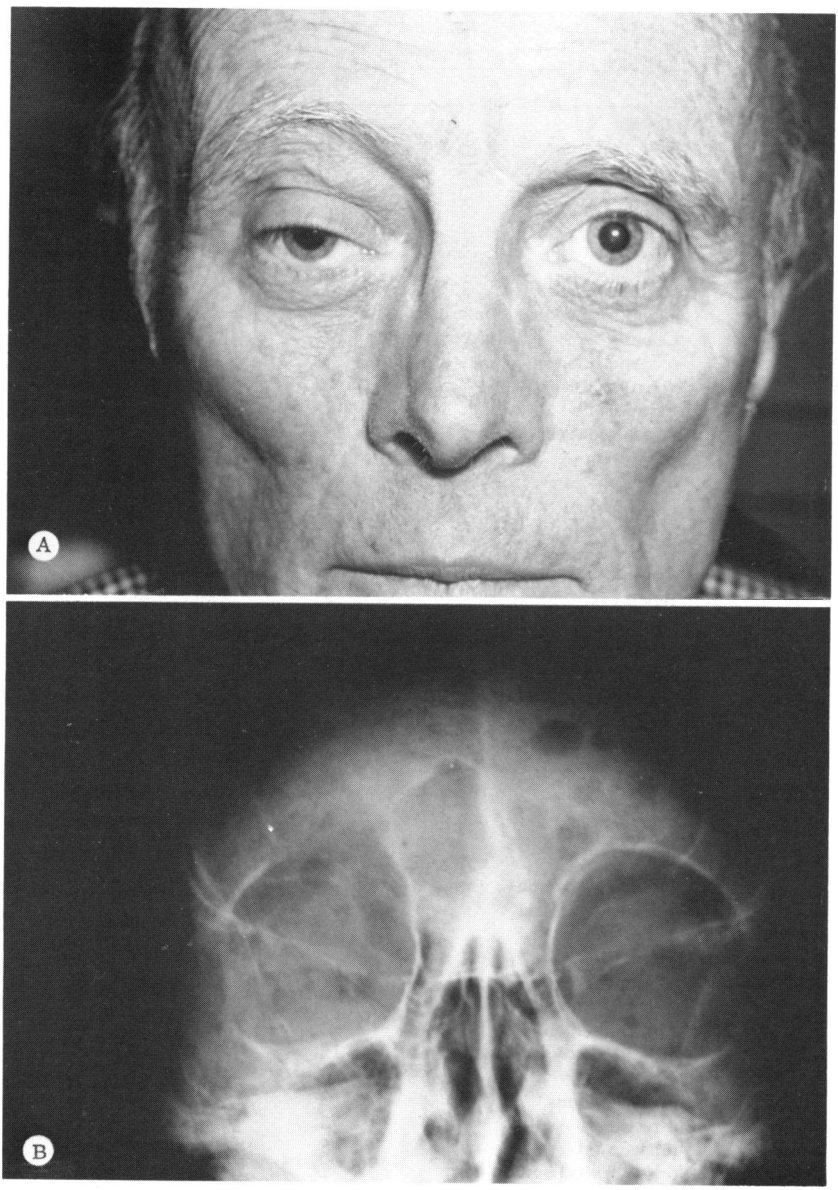

FIGURE 1

A: Seventy-year-old man with right proptosis and known multiple myeloma. Orbit not biopsied. B: Same patient showing Caldwell projection with punched out defects characteristic of multiple myeloma and right superior orbital rim involvement.

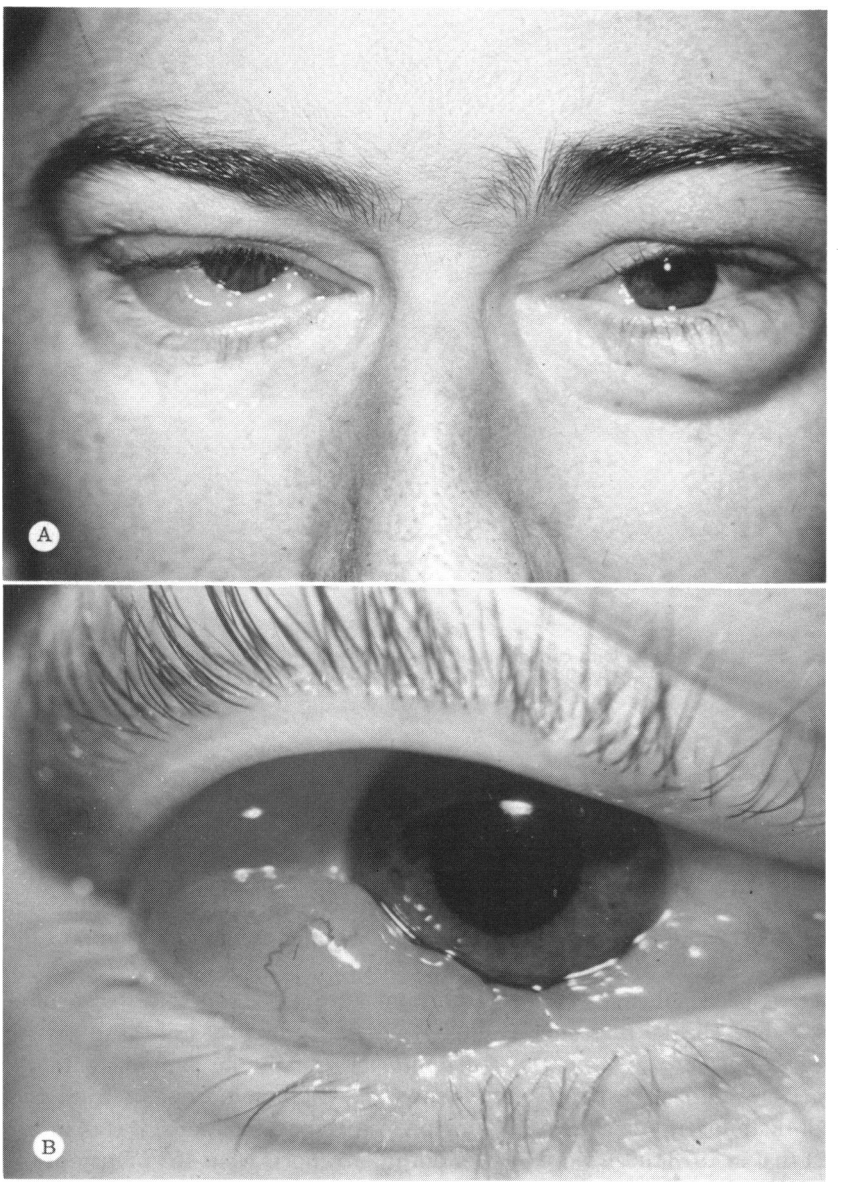

FIGURE 2
A: Hodgkins lymphosarcoma in 38-year-old man. Orbit not biopsied. B: Right eye, same patient.

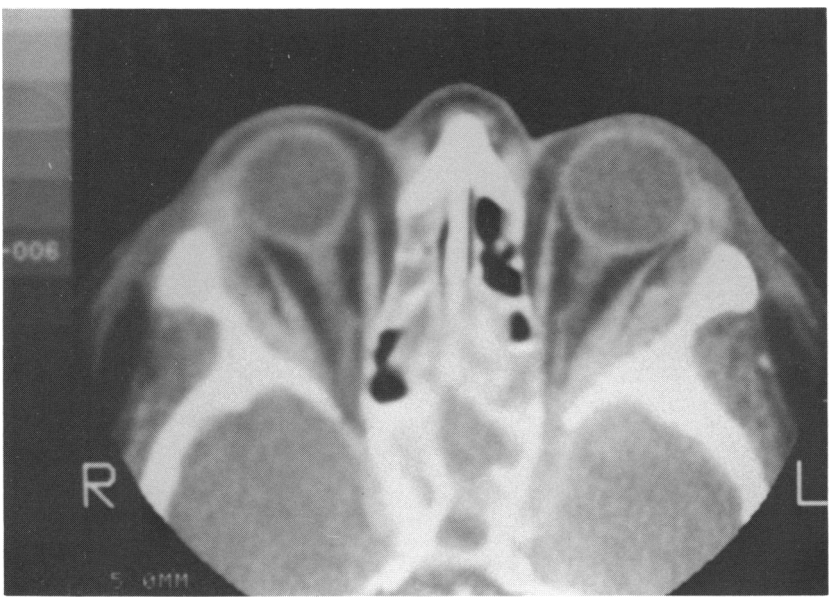

FIGURE 3
Sixty-five-year-old man with carcinoma of prostate, history of collapsed vertebrae, rib and clavicle involvement, markedly reduced vision due to bilateral orbital involvement demonstrated by CT scan.

Among the six cases reported in the present series there has been no recurrence or deaths to date. With only four to six to eight cases previously reported at the 40 to 50 medical centers, I feel it warrants pooling the available information for study and for prognosis on behalf of the patient. This emphasizes the importance of reports on biopsy-proven series of orbital lesions. This information can offer tremendous support for the clinician in the field.

Factual conclusions can be ascertained from a large series of orbital cases and special reference is made to certain categories of these cases.

Pseudotumor/inflammatory granuloma remains an enigma with its variable clinical response and the wide variation in reports from the ophthalmic pathologist. In Henderson's[18] two presentations, it is of interest that in 1973 there were 37 pseudotumor reports from 465 biopsy-proven orbital lesions for an incidence of 8%. In 1980[18] with the same lesions and additional cases presented for a total of 764 cases, at the 8% incidence, one would project 61 cases. In fact, there were fewer than in the first report, 35 or 4.6%. This attests to the changing field of ophthalmic

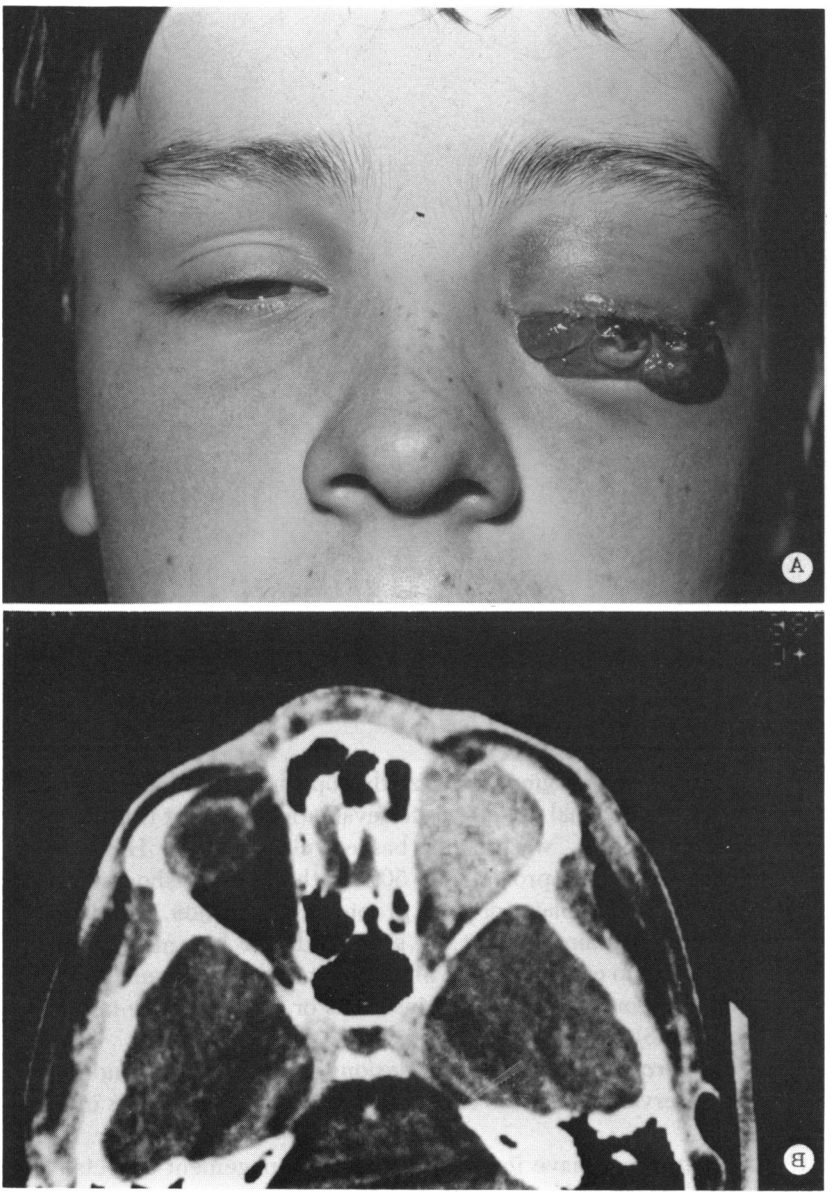

FIGURE 4

A: Fifteen-yar-old boy with basilar skull fracture and orbital hemorrhage with marked proptosis and corneal exposure. B: Same patient showiong extent of hemorrhage, evacuated temporally 2 weeks after injury with 20/50 visual residual due to corneal scarring.

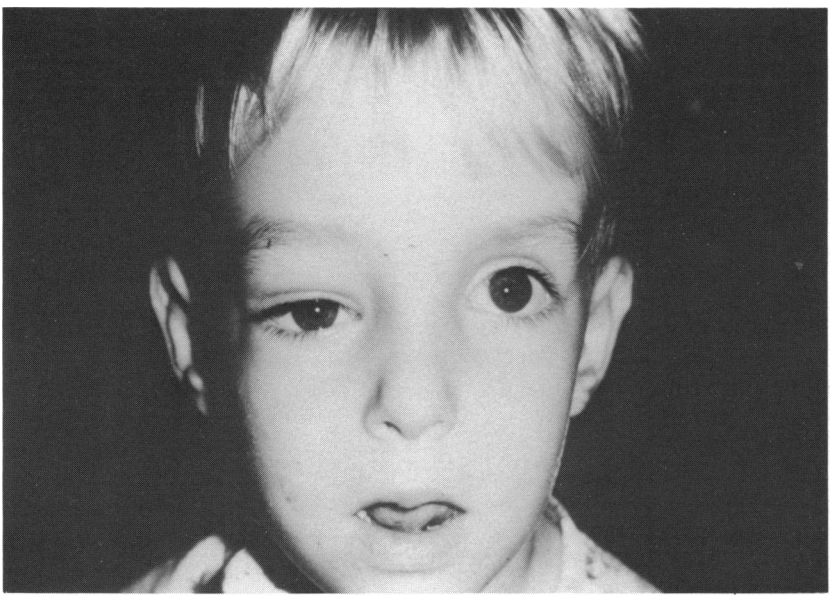

FIGURE 5
Three-year-old boy with fracture of roof of right orbit with progressive "growing fracture" or leptomeningeal cyst formation.

pathology and is encouraging to realize that ophthalmic pathology is more definitive with additional modalities for evaluation and that this diagnosis is becoming less of a "wastepaper basket" classification. Henderson's figures[18] demonstrate approximately 50% of the cases to show improvement in the initial complaint of pain, proptosis, congestion, and diplopia. Twenty percent remained about the same and 30% became worse with vision decreasing to complete loss. Six percent of the total were enucleated. The clinical course can be very insidious or have an acute inflammatory onset (Fig 8).

Basal cell carcinoma and other lid lesions warrant respect and careful continued observation because of their threat for orbit and sinus invasion (Fig 9).

Cystic lesions may have important clinical management aspects. This series had 45 *dermoid* lesions with two thirds being in the conventional upper temporal aspect of the orbit. Dermoid lesions were excluded unless retrobulbar or with bony defects. Five (11%) showed intracranial extension with one of these recurring, requiring a combined neurosurgi-

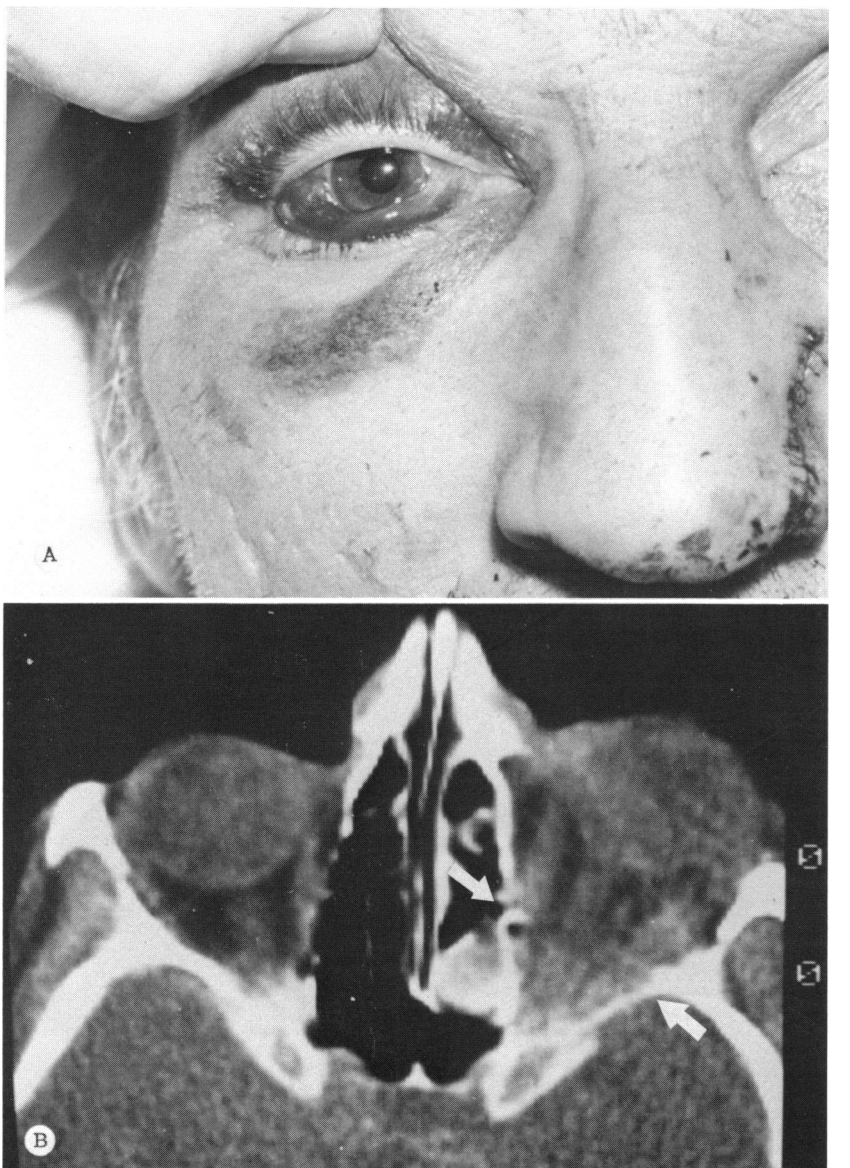

FIGURE 6
A: Forty-seven-year-old man with a stab wound left nares into right orbit severing optic nerve. B: CT of same patient. Optic nerve is seen to be severed. There was late manifestation of left middle fossa intracranial fluid warranting medical and neurosurgical supervision.

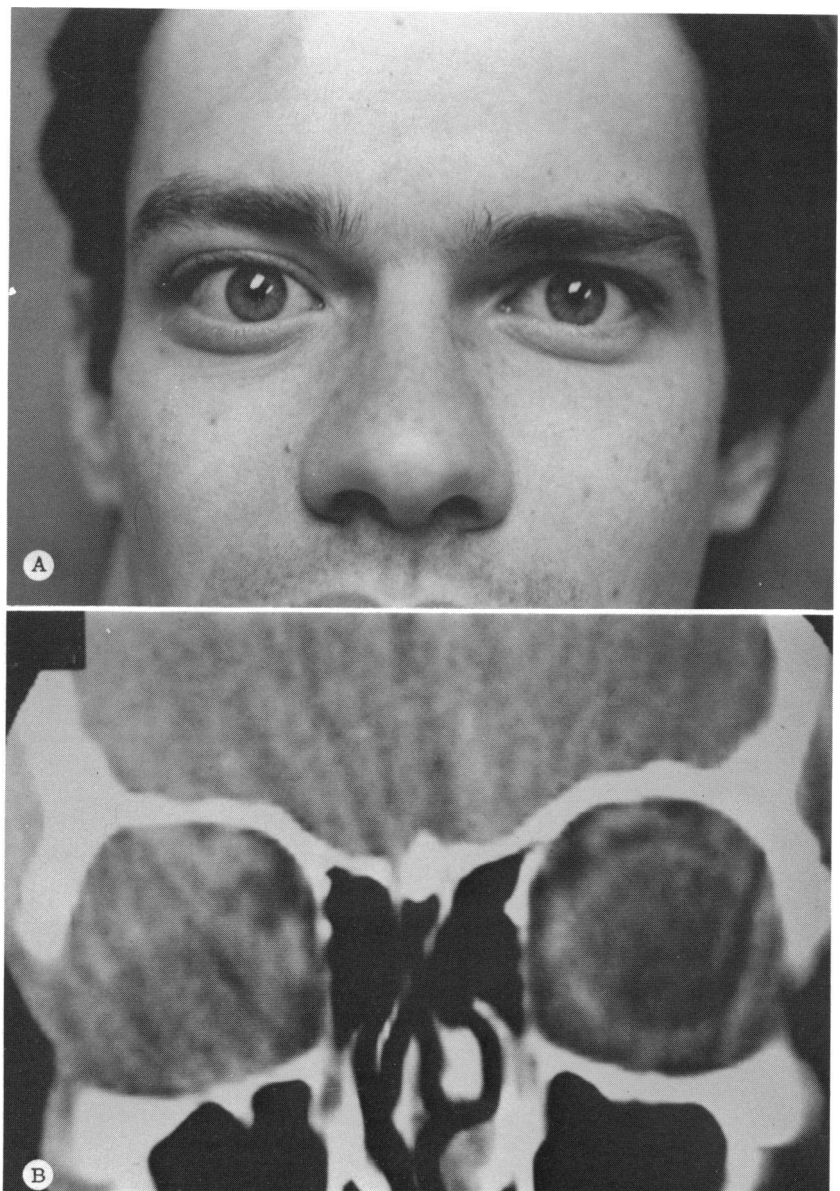

FIGURE 7
A: Twenty-eight-year-old man with proptosis OD, retinal striae and distorted vision. B: CT scan of same patient with large right orbital tumor and downward displacement of globe.

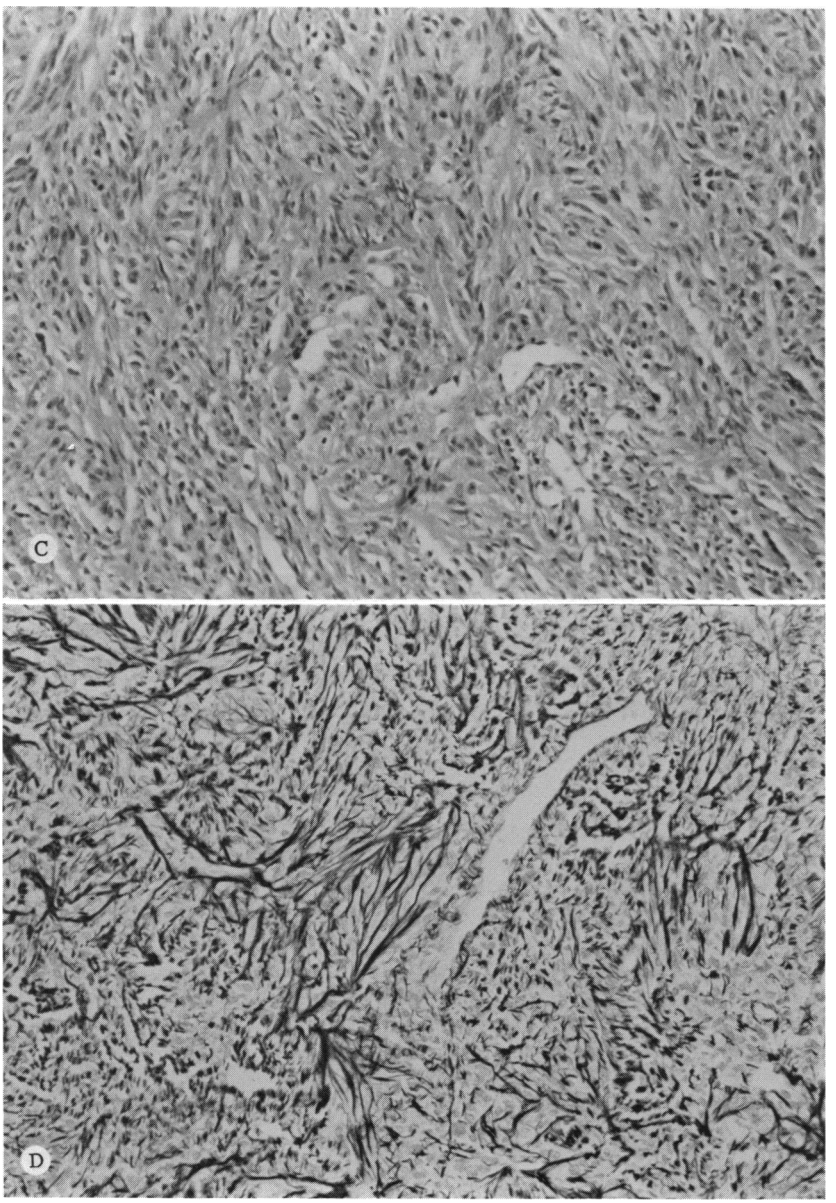

FIGURE 7 (Cont'd)
C: Pathology of same patient in A, showing apparent hemangiopericytoma (hematoxylin and eosin, original magnification, × 20). D: Same patient showing apparent hemangiopericytoma (reticulum stain, original magnification, × 20).

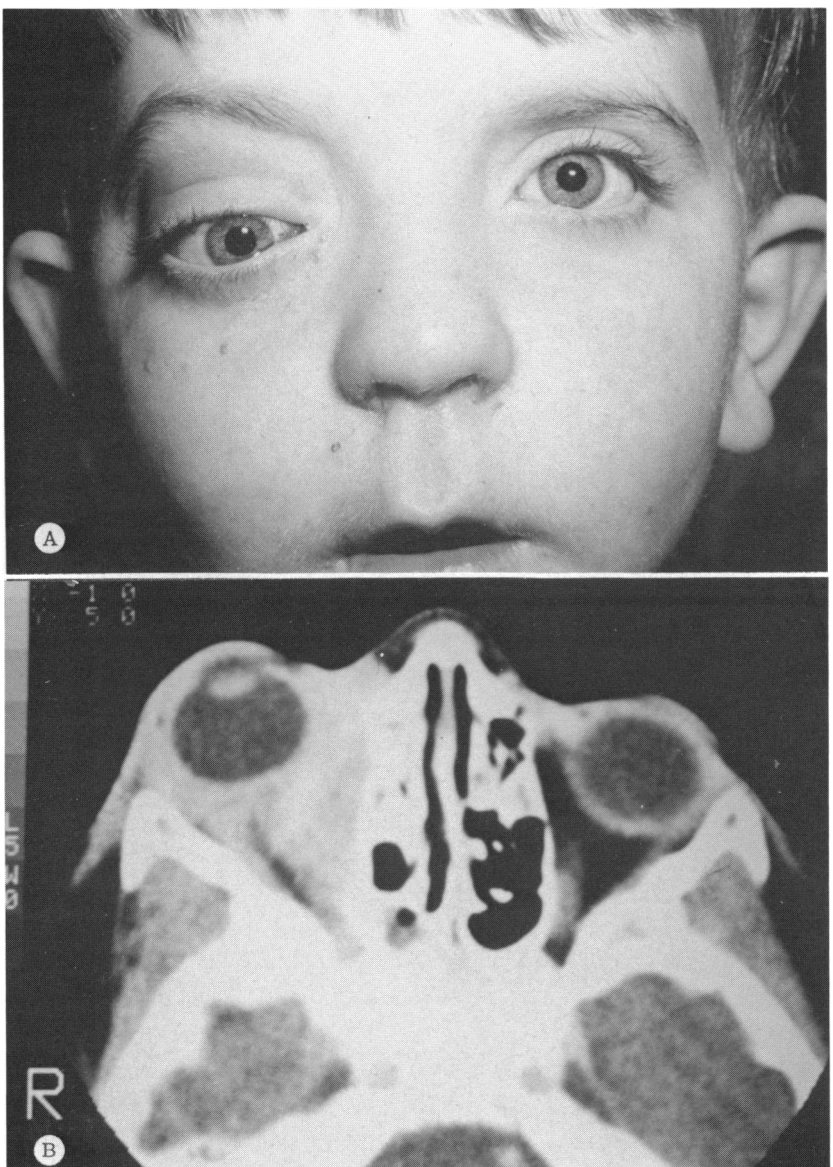

FIGURE 8
A: Five-year-old boy with rapid onset of proptosis OD and global displacement with intermittent exacerbations over 3 years despite steroids. B: CT scan showing extensive orbital involvement OD.

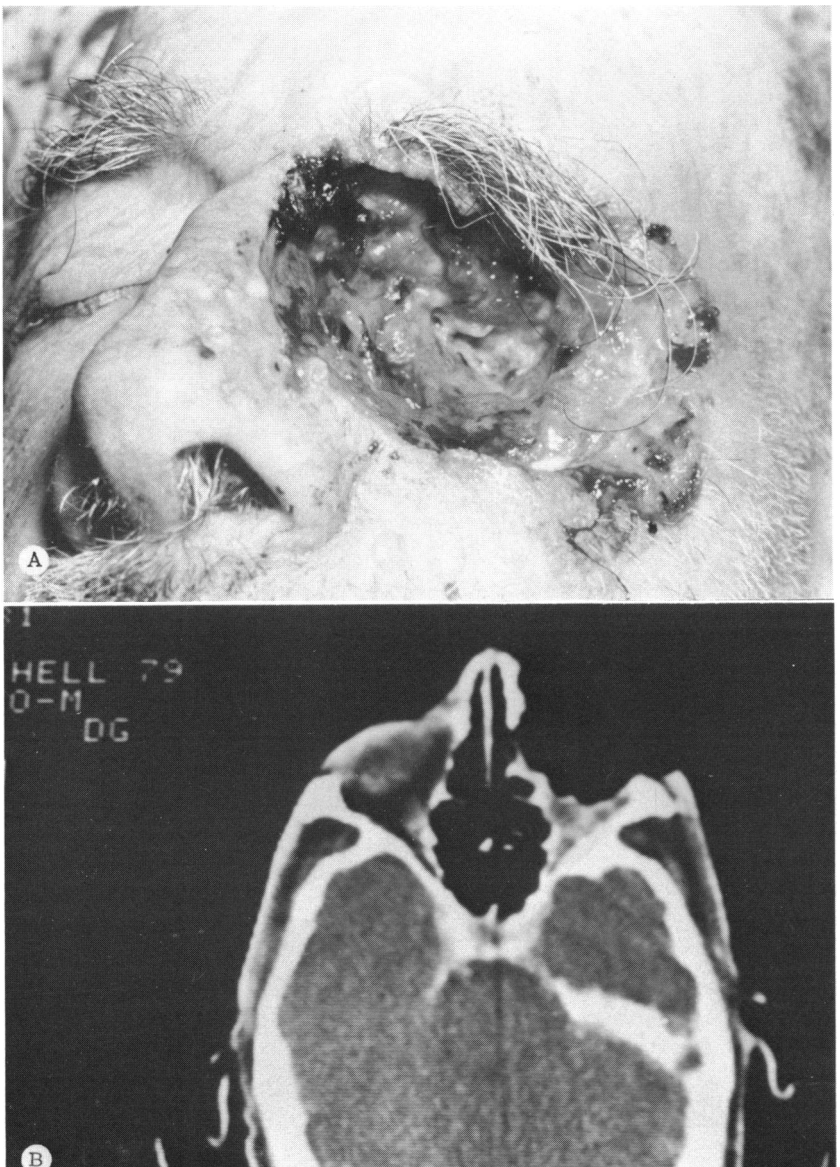

FIGURE 9
A: Seventy-nine-year-old man with basal cell carcinoma, left lower lid, who neglected repeated follow-up attempts, resulting in complete loss of the eye in a virtual "do-it-yourself" exenteration over a 15-year period. B: CT scan of same patient.

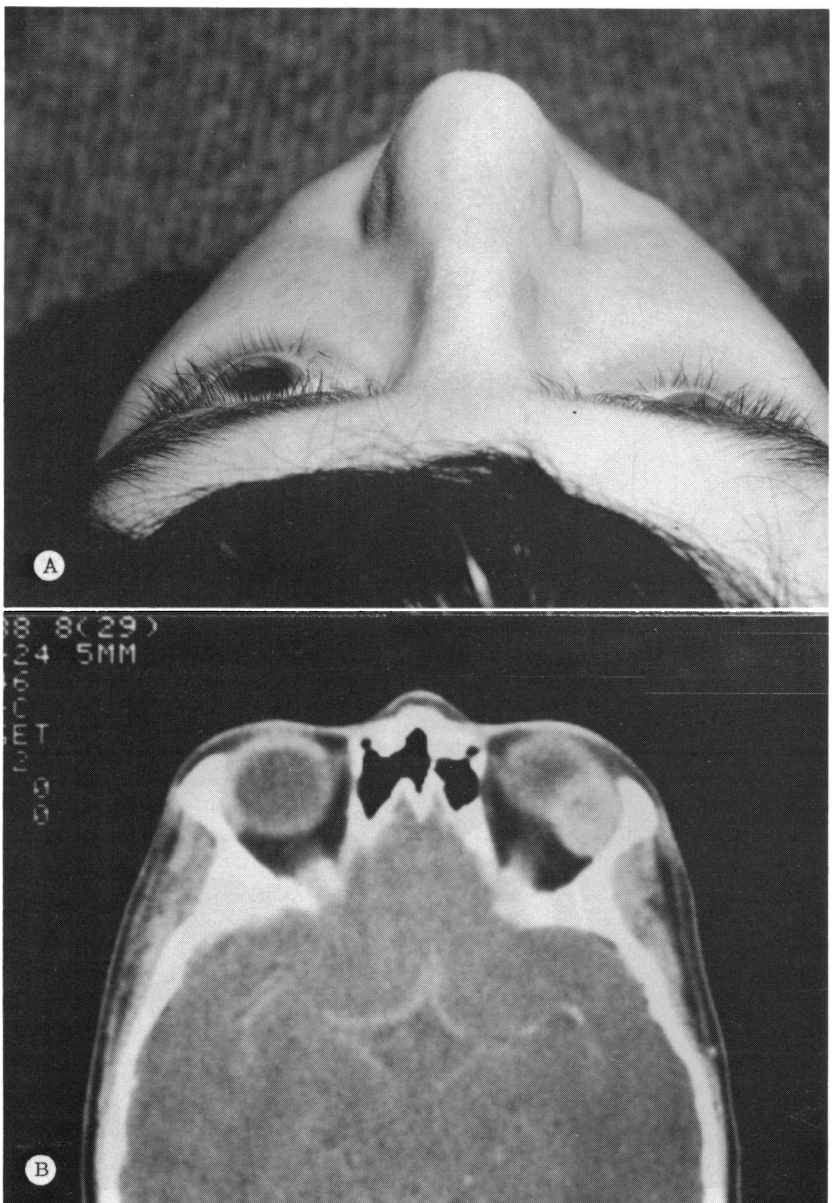

FIGURE 10
A: Twenty-nine-year-old woman with proptosis OS and diplopia. B: CT scan showing lacrimal fossa area tumor.

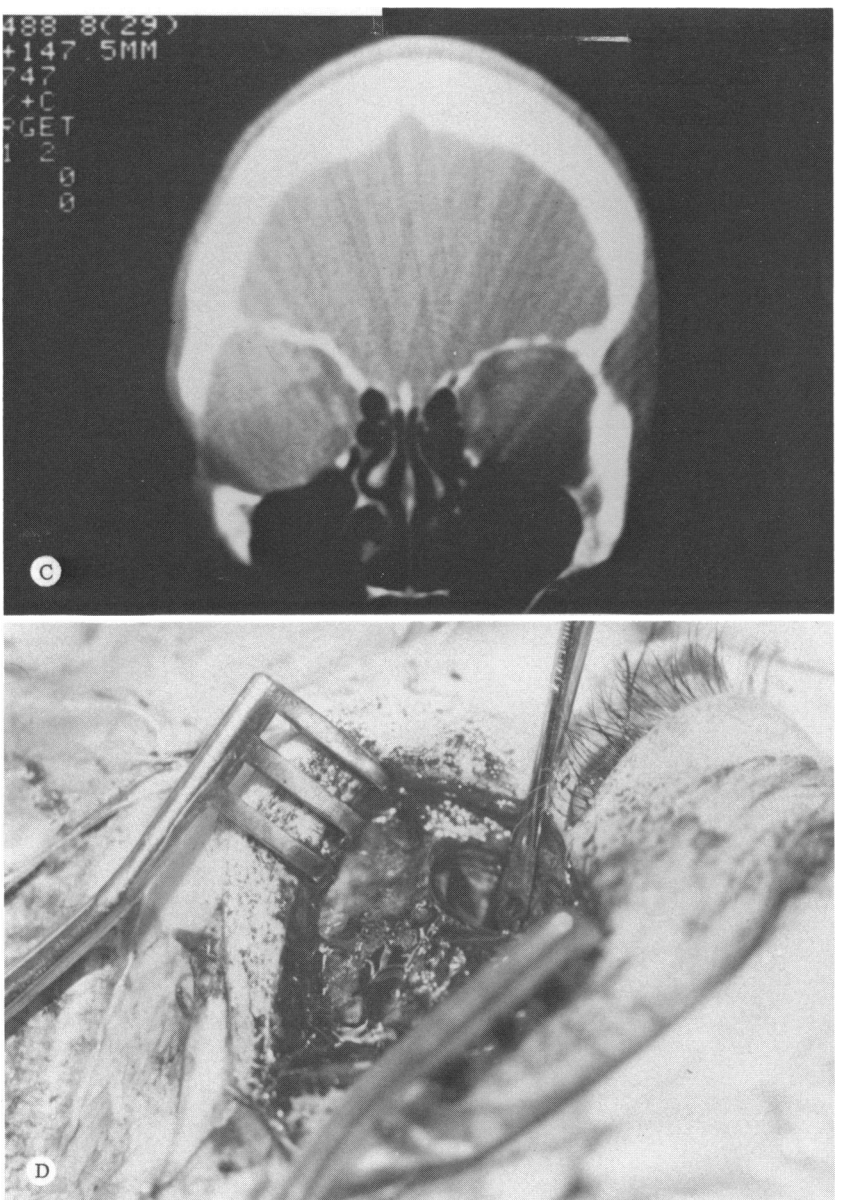

FIGURE 10 (Cont'd)
C: CT scan showing large lacrimal fossa area tumor. D: Same patient as in A, with skin incision leaving lateral canthus intact for better cosmetic result.

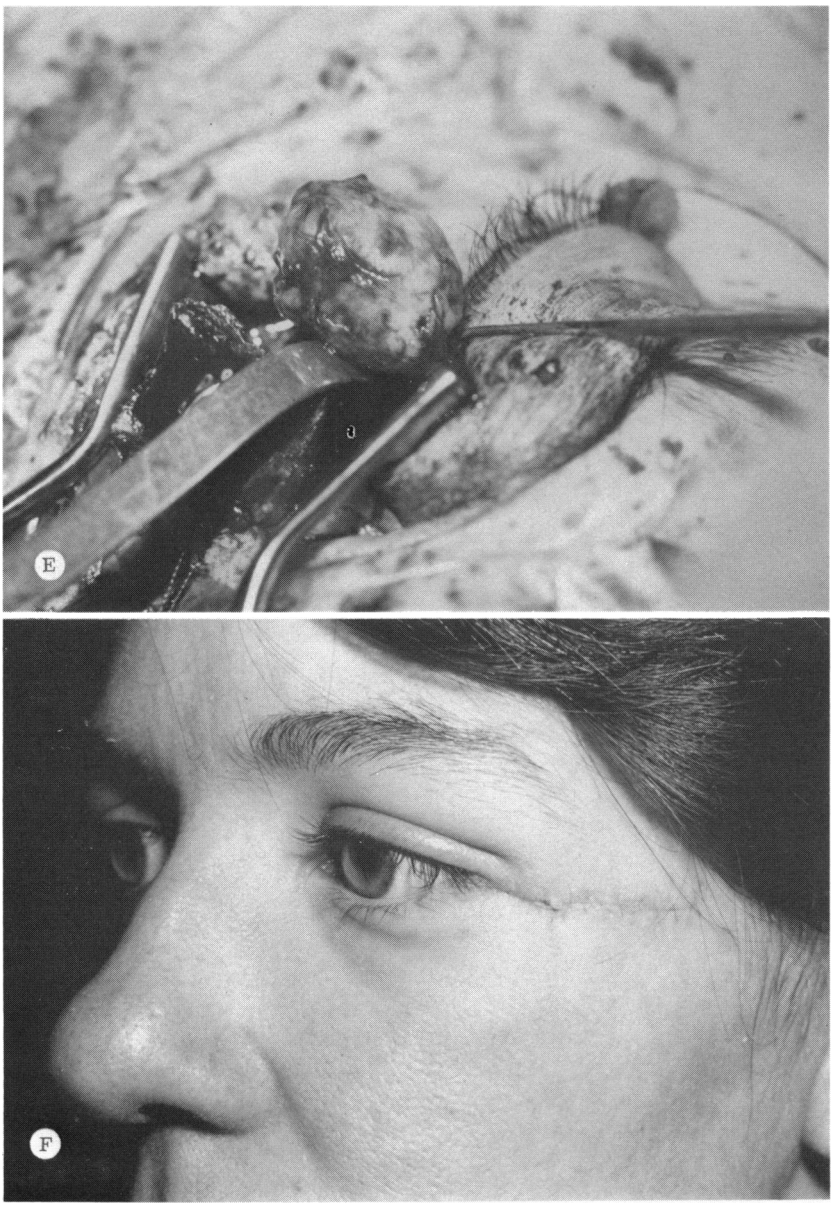

FIGURE 10 (Cont'd)
E: Lateral wall still attached to temporalis muscle but reflected/retracted for exposure. Exposure and removal of mixed cell tumor lacrimal gland. F: One week postoperative, skin suture in place, lateral canthus uninvolved, little reaction.

cal approach. Even though some had extensive bone involvement, this has been the only recurrence (2.2% incidence). Eight (18%) showed dural exposure at the time of surgery calling for particular care. Three (6.7%) extended through the lateral wall both in the orbit and temporalis fossa. With chewing action of the temporalis muscle, proptosis resulted which could be reduced by pressure on the globe, causing fullness in the temporalis fossa. Twenty-two (50%) required lateral wall removal for surgery.

Mucoceles number 31 in the series, ranging from 12 to 72 years of age. Dural exposure was noted in eight (26%) calling for care at the time of surgery. The Lynch procedure was effective in reestablishing drainage, with only one recurrence (3%) cured by a second procedure. Mucoceles were associated with fibrous dysplasia in three patients.

Multiple lesions may be present in a single patient. Meningiomas may be multicentric in approximately 14% of patients. One patient exhibited fibrous dysplasia with polyps operated and resulting in an orbital abscess requiring drainage, causing central artery occlusion, optic atrophy, and exotropia requiring surgery, followed by a mucocele requiring surgery, in addition to chronic sinus drainage procedures.

The malignancy rate for orbital lesions is of significance. In this clinical series of 820 cases, if the thyroid, trauma, fracture, congenital, and no diagnosis cases (187) are excluded, there are 633 orbital lesions. Two hundred fourteen, or 34% proved to be malignant. This warrants aggressive management rather than a casual watchful waiting supervision of these patients.

Of the 820 clinical orbital cases, over 500 were operated on with 450 biopsy proven. Most of the others were traumatic cases.

Surgical Management. With improved techniques, surgery has progressed to the point of being performed earlier and with more precision than ever before. Our ability has been aided by experience acquired over the past 30 to 40 years, improvement in radiologic and associated tests, ultrasonography, and the phenomenal improvement in CT. The axial and coronal projections give a three-dimensional aspect to the surgical approach and reduce "hunting for a needle in the haystack" to pinpoint accuracy in the approach to the known tumor location.

The surgical incision for bone removal usually spares the lateral canthus for a better cosmetic result (Fig 10). The lateral wall frequently can be rotated temporally and left attached to the temporalis muscle which allows good repositioning and eliminates the necessity for wiring. A full lateral wall approach is best for a lacrimal fossa area lesion for good exposure to attempt to remove a lacrimal gland tumor intact. A "small biopsy" through a skin incision may only disrupt a lacrimal tumor and

allow possible seeding for regrowth or spread. It may also give a false sense of security when a small area of normal gland is removed, thus giving a false reading, when in fact it has been pushed forward by an actual tumor.

Invariably, a drain is not necessary except with a known orbital abscess from sinusitis, uncontrolled bleeding, or a ruptured deep dermoid because of the marked reaction to the cyst contents. Antibiotics are rarely used except with known infection.

Lid suturing is seldom necessary unless there is marked proptosis. Vision should be recorded and the dressing changed. It is rare for the patient to be hospitalized more than 2 days even after a lateral wall approach.

For the pathologist, a frozen section of the orbit is encountred infrequently and often cannot be definitely interpreted. Many must be read from regular sections or even require a consultation opinion. I have had to wait 2 and even as long as 4 weeks for a definitive decision. It is well to advise the patient of this delay before surgery. Extensive or radical surgery should only be carried out with a positive frozen section reading because of the difficulty in reading lymphocytic reaction of inflammation versus malignancy.

SUMMARY

Eight hundred twenty clinical orbital cases have been reviewed as seen over a 34-year period. These are tabulated as to classification, numbers, and percentages. They include biopsy-proven (450 cases) and other clinical cases. These are patients seen by ophthalmologists practicing in an average community area rather than a large referral center and thus may be more truly representative of what might be encountered in practice.

Any series has a certain bias. The advantage of a biopsy-proven series is recognized for the certainty and prognostic aspect. A clinical series represents a wider practical correlation and may include cases not likely to be biopsied, such as hemangioma. It is hoped that the percentage figures of these types of cases will help the clinician in his diagnostic evaluation of patients with orbital lesions.

In this series 34% of the orbital lesions were malignant which warrants an aggressive approach for such cases rather than conservative supervision.

Because of the very limited number or absence of some types of cases even in a large clinical or biopsy-proven series, I feel it warrants the development of a standard classification of lesions to be used by everyone.

Also, assembling the data on infrequent types of lesions from a number of medical centers would give a much larger series and would have a more meaningful prognostic significance. This might be accomplished through one of our national ophthalmologic societies.

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DISCUSSION

DR IRA JONES. Since the number of orbital cases is small and is dispersed in many hands, the addition of a large series handled by one capable and experienced orbital surgeon is a noteworthy event. These cases, collected over 34 years and representing referrals from about 50 ophthalmologists in the same geographic area, are representative of the spectrum of cases which might be encountered by the general ophthalmologist. Although Doctor Kennedy suggests that the varieties and numbers will be different from those encountered in a large referral practice in a medical center, such does not prove to be the case when compared with the cases collected over the past 50 years at the Edward S. Harkness Eye Institute in New York. His percentage of malignant tumors is 34 and ours is 32. All of the other parameters are likewise, fairly equal. A surprising aspect of this study to me is that the number of orbital cases in his area seems to be increasing, whereas the numbers in several other centers with which I am familiar, are decreasing. There seems to be two reasons for the decrease in other centers. One is that the number of trained orbital surgeons has increased so that the backlog of cases has been reduced. The chief reason, however, is the improvement in diagnostic skills brought about by the combined use of orbital ultrasound measurements, axial and coronal computed tomograms, and the emerging technique of nuclear magnetic resonance. If a reasonably satisfactory diagnosis can be arrived at without invasive techniques, this eliminates the need for a dangerous and possibly harmful procedure. I understand that the scope of this paper had to be restricted, but I was nevertheless disappointed not to hear the problem of the complications of orbital surgery addressed. I hope Doctor Kennedy in a future paper will share with us his surgical techniques, his complications, and the measures which he employed to avoid complications.

DR ROBERT KENNEDY. I would like to thank Doctor Ira Jones for being the primary discussant. He certainly was most gracious with his comments. I consider him one of my mentors who has frequently offered advice and guidance. Fortunately, he is only 57 air-minutes away from many patients who can easily fly to New York City for consultation, advice, or management.

It is of interest that the variety and percentage numbers encountered in this series, and the percentage presence of malignant tumors are all quite similar to the much larger series as handled over 50 years at the Harkness Eye Institute. The one in three malignancy rate is significant.

This series is not made up of consecutive cases. This is due to the fact that while working, assembling pictures and office records of a series of these cases in my home, a plug-in flashlight that was being recharged got so excited it burst into flames when the football player, O. J. Simpson, ran for a record 2003 yards that season. A large number of slides and records were destroyed by fire. Thus it has taken awhile to build back up to the numbers presented today.

Quite likely this paper is too long. As Doctor Jones suggests, the problems and complications of orbital surgery, surgical techniques, and measures employed to avoid complications, is another whole area of interest which might be addressed at another time. I appreciate the opportunity to have presented this material.